VOLUME - 11, ISSUE - 10, OCTOBER - 2022 • PRINT ISSN No. 2277 - 8160 • DOI : 10.36106/gjra

Original Research Paper

Histopathology

ANGIOLYMPHOID HYPERPLASIA WITH EOSINOPHILIA – A RARE BENIGN VASCULAR TUMOR

Dr. Kinjal Tandel	3rd year Resident Doctor, Pathology Department
Dr. Alaukika Rathwa	Associate Professor, Pathology Department
Dr. Apeksha Malani*	3rd year Resident Doctor, Pathology Department *Corresponding Author

ABSTRACT **Background:** Angiolymphoid hyperplasia with eosinophilia is a rare benign vascular tumor of uncertain etiology. It is most commonly seen in middle aged females and presented with single or multiple nodules in head and neck region. **Case report:** A 26-year female patient presented with multiple nodules over right cheek and mandibular region and history of right cheek swelling. On examination swelling was single, mobile, non-tender and well-defined present over right cheek and violaceous pedunculated multiple papules and nodules were present over right jaw. CBC was within normal range with normal eosinophil count and MRI showed possible neoplastic lesion in neck region. Histopathological features were suggestive of Angiolymphoid Hyperplasia with Eosinophilia. **Conclusion:** Even though Angiolymphoid hyperplasia with eosinophilia is a rare entity, it has to be differentiated from Kimura's disease as they have different therapeutic implications.

KEYWORDS : Angiolymphoid hyperplasia with eosinophilia, middle age female, Kimura's disease

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia, term was introduced first by Wells and Whimster in 1969[1]. It is a rare benign vascular tumor of uncertain etiology. Various other names are Epithelioid hemangioma, Inflammatory angiomatous nodule, Papular angioplasia and Intravenous atypical vascular proliferation[2]. It is most commonly seen in middle aged females and presented with single or multiple nodules in head and neck region[3].

Case Study

A 26-year female patient presented to skin OPD with multiple nodules over right cheek and mandibular region since 3 months and history of right cheek swelling since 6 months. Patient was referred to ENT OPD for right cheek swelling.

On examination:

1. Single, mobile, nontender well defined 3x3 cm size swelling present over right side of cheek.
2. Violaceous pedunculated multiple papules and nodules present over right jaw (figure 1).

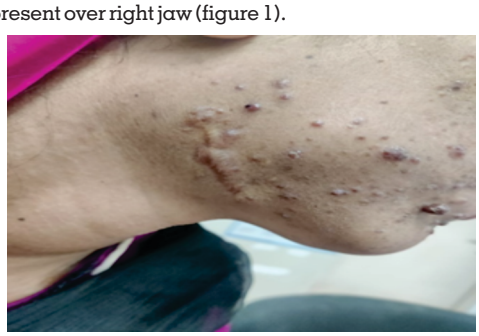

Figure 1: Swelling over right cheek and violaceous pedunculated multiple papules and nodules present over right jaw (Figure 1).

Investigations:

CBC report was normal and eosinophil count was within normal range.

USG local part (Right cheek):

Evidence of 33x26x15 mm sized well defined hypoechoic lesion at right side of cheek with internal vascularity seen within.

Multiple enlarged matted lymph nodes are seen at right submandibular, largest measures 46x17 mm.

MRI neck with oral cavity:

Abnormal signal intensity lesion in the region of the right cheek and right buccal mucosa, opposite the right lower 2nd and last molar teeth, loss of plane between lesion and the anteroinferior aspect of right masseter muscle. The lesion is abutting adjacent body of the mandible on right side, however no obvious abnormal signal or bone erosion noted. Enlarged lymph nodes in the right submandibular and upper internal jugular regions and bilateral submental regions.
Possible neoplastic lesion.

Intervention:

Cheek swelling was excised and sent to histopathology department.

Pathological Findings

Single, whitish, globular measuring 3.5x2x1 cm tissue mass was received (Figure 2).

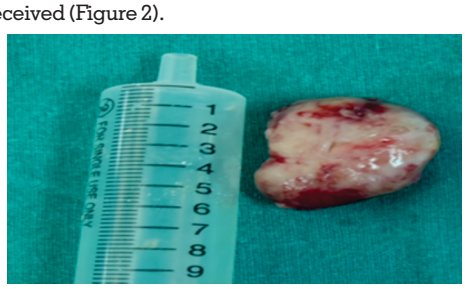

Figure 2: Single, whitish and globular tissue mass.

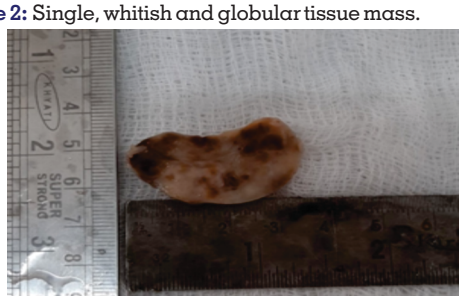

Figure 3: On cut surface: multiple small hemorrhagic areas present.

Microscopically, proliferation of vascular channels with nest and cords of endothelial cell proliferation surrounded by severe inflammatory cells infiltrate consisting of eosinophils, plasma cells and lymphocytes with germinal center formation.

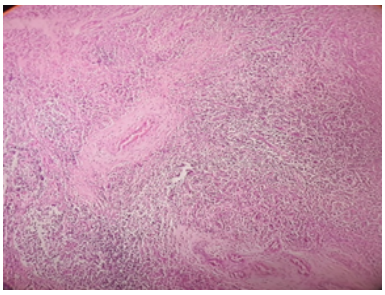

Figure 3: Proliferation of vascular channels with nest and cords of endothelial cell

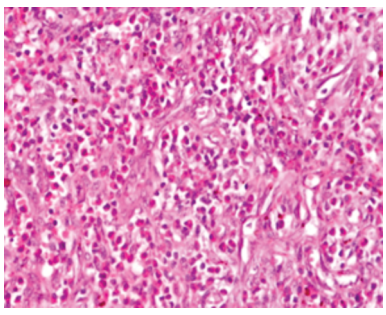

Figure 4: Severe inflammatory cells infiltrate consisting of eosinophils, plasma cells and lymphocytes

DISCUSSION

Angiolymphoid hyperplasia with eosinophilia (ALHE) was described by Wells and Whimster in 1969[1]. It is characterized by proliferation of blood vessels lined by endothelial cells and surrounded by inflammatory infiltrate comprising of eosinophils, mast cells and lymphocytes.

Etiopathogenesis of ALHE is unknown. Some hypotheses such as a reactive process, a neoplastic process and infectious mechanism with possible association with HIV are explained[4]. Most common sites of ALHE are peri-auricular region, forehead and scalp. Other sites are orbit, oral mucosa, shoulders, hands, breast, liver, spleen, heart, bone and penis[5,6].

Angiolymphoid hyperplasia with eosinophilia is a distinct entity from Kimura's disease. Kimura's disease mainly seen in young Asian men with one or multiple asymptomatic masses involving subcutaneous tissue and salivary glands. Microscopically, Kimura's disease presents numerous lymphoid follicle formation in the deeper part, dense fibrosis and eosinophil microabscess[6].

CONCLUSIONS

Even though Angiolymphoid hyperplasia with eosinophilia is a rare entity, it has to be differentiated from Kimura's disease as they have different therapeutic implications. On the basis of clinical and histopathological presentation, we came to final diagnosis of angiolymphoid hyperplasia with eosinophilia in this case report.

REFERENCES

1. Wells GC, Whimster IW. Subcutaneous angiolymphoid hyperplasia with eosinophilia. *Br J Dermatol*. 1969;81(1):1–5.doi:10.1111/j.13652133.1969.tb15914.x.
2. Park Y, Chung J, Chang-Gun C. Angiolymphoid hyperplasia with eosinophilia of the tongue: report of a case and review of the literature. 2020;5(4):317–319 319 Oral Oncol. 2002;38(1):103–6. doi:10.1016/s1368-8375(01)00020-3.
3. Quattrocchi CM, Jankovic R, Jacquier M, Sánchez A, Bergero A, Bussy R, et al. Hiperplasia angiolinfoide con eosinofilia: reporte de un caso. *Arch Argent Dermatol*. 2012;62:189–92. doi:10.1046/j.13652133.2001.04364.x.
4. Guo R, Gavino AC. Angiolymphoid hyperplasia with eosinophilia. Arch Pathol Lab Med. 2015;139(5):683–6.
5. Meyerle J, Glusac E. Angiolymphoid hyperplasia with eosinophilia 2005. Available from:
6. http://www.emedicine.com/derm/topic30.htm [Last accessed on 2006 Jan]. de Freire Cassia F, Avelleira JCR, et al. Angiolymphoid hyperplasia with eosinophilia versus Kimura's disease: a case report and a clinical and histopathological comparison. *Anais Bras de Dermatol*. 2017;92(3):392–4. doi:10.1590/abd1806-841.20175318.